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Catastrophic intraoperative bleeding due to congenital extrahepatic porto-systemic shunt anomaly: A surgical case report of two rare anomalies

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ABSTRACT

INTRODUCTION: Abernethy malformations are extremely rare congenital anomalous portosystemic shunts. We report the case of a patient with a rare variant Abernethy malformation between the superior mesenteric vein and left renal vein, associated with a massive jejunal diverticulum.
PRESENTATION OF CASE: A 37-year-old Caucasian female presented to our emergency department with severe abdominal pain and proceeded to laparotomy for a presumed small bowel obstruction. At laparotomy she was found to have a massive diverticulum at the duodeno-jejunal junction, which was intimately associated with a venous malformation and the anomalous portosystemic shunt. Whilst mobilising the diverticulum, the patient developed catastrophic haemorrhage from the malformation. The patient underwent a complicated post-operative course however was eventually stabilised.
DISCUSSION: We discuss the anatomy and pathophysiology of anomalous portosystemic shunts and propose an embryological origin for our patients' anomalies.
CONCLUSION: Abernethy malformations are rare however may be associated with other intra-abdominal pathology and extreme caution is required when operating on these patients.
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1. Introduction

Congenital Extrahepatic Porto-Systemic Shunts (CEPS) were first described by Abernethy in 1793 discovered at autopsy of a female infant. The majority of reported cases have been discovered in childhood with a greater prevalence in females[1], although they can seemingly present in any age group.

Morgan and Superina [2] categorised CEPS into two anatomical classes based on the absence or persistence of hepatic portal venous flow. Type 1 is a complete shunt, with no portal venous blood able to enter the hepatic circulation. Type 2 is a partial shunt, in which an accessory venous pathway from the portal to the systemic venous system is present in addition to the portal vein. In Type 2 anomalies the portal vein is often hypoplastic. Kobayashi et al. [2] has further classified CEPS by the destination of their outflow, with the most common outflow being into the inferior vena cava (IVC) and designated Type A, being distinguished from those with outflow into the renal veins (Type B) and the iliac veins (Type C) (Fig. 1).

CEPS may be asymptomatic or may lead to a wide range of sequelae. They may be an isolated pathology but are more often associated with other anatomical anomalies [3,4].

We report the case of a patient with a rare Abernethy malformation between the superior mesenteric vein (SMV) and left renal vein (LRV) associated with a massive jejunal diverticulum.

Our case report work has been reported in line with the SCARE criteria [5].

2. Presentation of case

A 37-year-old woman presented to the emergency department with 12 h of severe epigastric abdominal pain. She had a background of un-investigated recurrent episodes of intermittent severe abdominal pain and had undergone a laparotomy as a 1-day old baby for an "abdominal cyst", the details of which were not available.

On examination, she was very distressed, she had a low-grade fever (38 °C), generalised abdominal tenderness with guarding in the epigastrium, and no features of chronic liver disease. She had a mild leucocytosis, and other blood tests including liver function tests were within normal limits.

A computed tomography (CT) of the abdomen and pelvis revealed what was presumed to be a dilated small bowel loop in the upper abdomen, 10 cm in diameter and containing faeculent mate-

Abbreviations: CEPS, anomalous extrahepatic porto-systemic shunt(s); CT, computed tomography; SMV, superior mesenteric vein; LRV, left renal vein; DJ, duodeno-jejunal.

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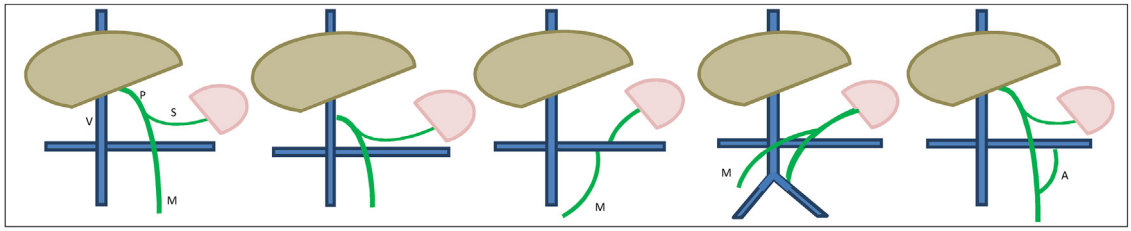

Morgan [3]	Normal anatomy	1	1	1	2
Kobayashi [2]	Normal anatomy	A	B	C	B

V= Inferior vena cava, P= Portal vein, S= Splenic vein, M= Superior mesenteric vein, A= Accessory shunt vessel

Fig. 1. Schematic examples of Abernethy malformation under the classification systems of Morgan et al. and Kobayashi et al.

Morgan [3]	Normal anatomy	1	1	1	2
Kobayashi [2]	Normal anatomy	A	B	C	B

V = Inferior vena cava, P = Portal vein, S = Splenic vein, M = Superior mesenteric vein, A = Accessory shunt vessel.

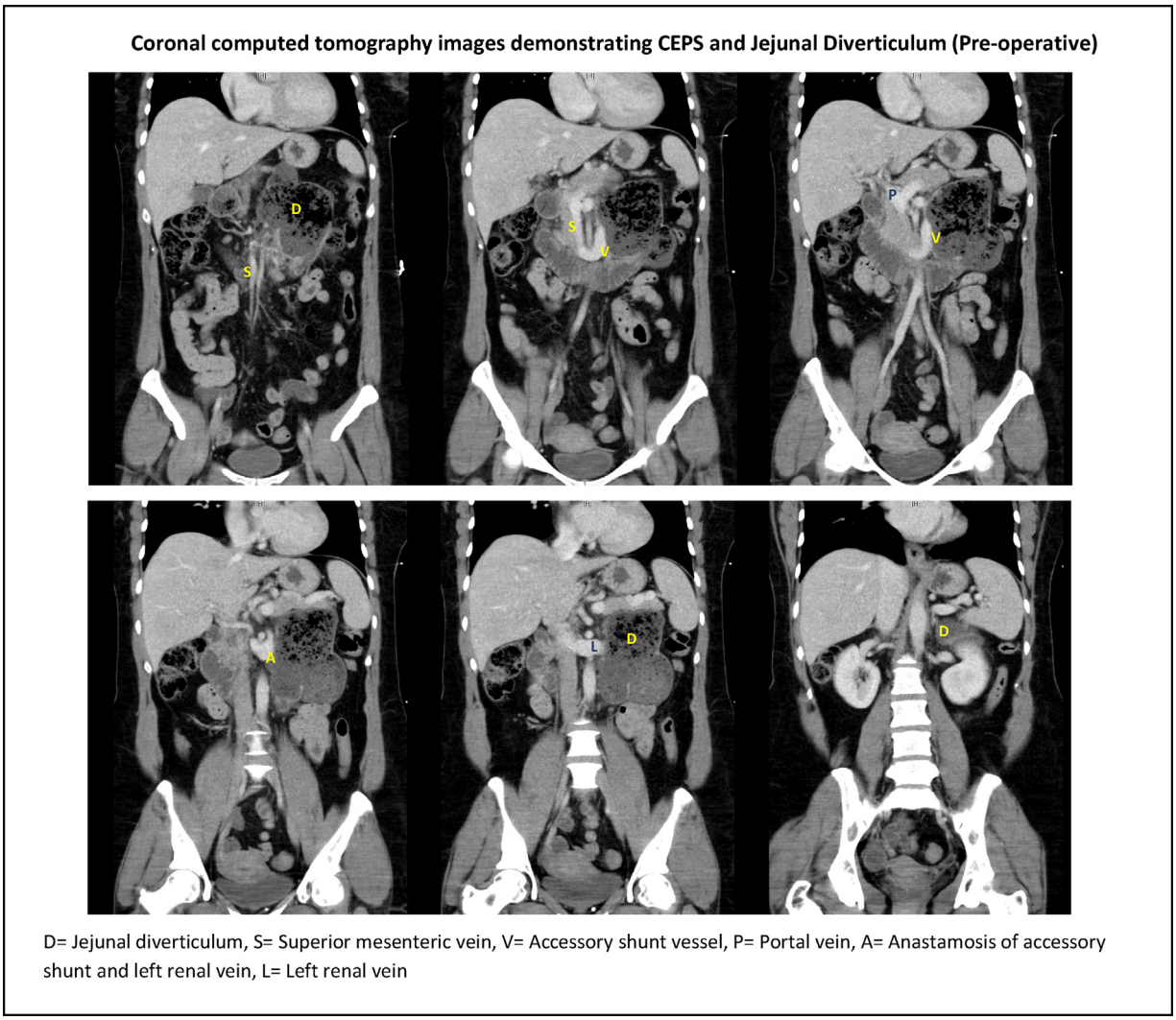

Fig. 2. Coronal computed tomography images demonstrating CEPS and Jejunal Diverticulum (Pre-operative).
D = Jejunal diverticulum, S = Superior mesenteric vein, V = Accessory shunt vessel, P = Portal vein, A = Anastamosis of accessory shunt and left renal vein, L = Left renal vein

rial. Considered differentials included a closed loop small bowel obstruction or abnormally located caecal volvulus. Furthermore, an abnormal vein was noted joining the superior mesenteric vein (SMV) to the left renal vein (LRV). This large vein originated as a side branch of the SMV at spinal level L2 and made a small inferior loop before ascending to spinal level L1 where it turned posteriorly to join the anterior aspect of the LRV in an end-to-side anastomosis (Figs. 2 and 3).

Despite large doses of opioid analgesia, the patient's pain remained severe and it was decided to proceed to laparotomy. At laparotomy adhesive bands due to previous laparotomy were found and single massive thin walled diverticulum was found at

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